



Original communication

A re-audit of the use of definitions of sudden infant death syndrome (SIDS) in peer-reviewed literature

Roger W. Byard MBBS, MD, Professor ^{a,b,*}, Vivian Lee Medical student ^{a,b}^a Discipline of Anatomy and Pathology, The University of Adelaide, Level 3 Medical School North Building, Frome Road, Adelaide, SA 5005, Australia^b Forensic Science SA, 21 Divett Place, Adelaide, SA 5000, Australia

ARTICLE INFO

Article history:

Received 11 January 2012

Received in revised form

13 February 2012

Accepted 8 April 2012

Available online 27 April 2012

Keywords:

SIDS

Infant death

Definition

Research

Audit

ABSTRACT

The use of different definitions of sudden infant death syndrome (SIDS) may make comparison of data among studies difficult. Fifty randomly selected papers dealing with SIDS that were published between 2010 and 2011 in peer-reviewed journals were reviewed to determine whether one of three internationally accepted definitions of SIDS had been either written in the text or referenced. A significant improvement in the use of definitions has occurred since 2005, with the percentage of papers either quoting or referencing a standard definition increasing by 26%, from 42 to 68%. The 1989 NICHHD definition remained the most commonly used definition (35.1%) followed by the 2004 San Diego definition (26.3%). Although the percentage of papers where either no definition was provided or where an idiosyncratic or mis-cited definition was used fell 26%, from 58 to 32%, nearly one in three papers published on SIDS in peer-reviewed journals that were included in this study still did not cite a standard definition.

© 2012 Elsevier Ltd and Faculty of Forensic and Legal Medicine. All rights reserved.

1. Introduction

In 2006, a study was conducted by one of the authors (RWB) to determine how often and how rigorously definitions of SIDS were being applied in peer-reviewed publications. Significantly, 58% of reviewed papers either used a non-standard or idiosyncratic definition, or did not specify a definition.¹ Given that considerable funding is provided for SIDS research and that conclusions from such investigations often have significant impact on families and their communities, it is important that the highest level of accuracy is maintained in defining cases and controls. It is recognised that when currently accepted international definitions of SIDS are adhered to, cases tend to be better investigated with the identification of a range of occult diseases and conditions.² To re-examine this issue and to determine whether there have been any changes in the intervening five years, a follow-up study was undertaken.

2. Materials and methods

As in the previous study, the United States National Library of Medicine 'Entrez PubMed' database³ was searched for all entries

listed under "sudden infant death syndrome" for the years 2010 and 2011. Of the 477 papers listed for 2010 to 2011, 50 were randomly selected from the printed abstracts where the conclusions depended on accurately defining SIDS. 'Letters to the Editor', 'Brief Reports' and commentaries that did not specifically focus on SIDS were excluded. The papers were then reviewed and checked to determine whether one of three internationally accepted definitions of SIDS^{4–6} had been either written in the text or referenced. The three definitions were:

- 1) *The Seattle definition* (1969): 'the sudden death of any infant or young child which is unexpected by history, and in which a thorough post-mortem examination fails to demonstrate an adequate cause of death.'⁴
- 2) *The NICHHD definition* (1989): 'the sudden death of an infant under one year of age, which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history.'⁵
- 3) *The San Diego definition* (2004): 'the sudden and unexpected death of an infant under 1 year of age, with onset of the lethal episode apparently occurring during sleep, that remains unexplained after a thorough investigation including performance of a complete autopsy, and review of the circumstances of death and the clinical history.'⁶

* Corresponding author. Discipline of Anatomy and Pathology, Level 3 Medical School North Building, The University of Adelaide, Frome Road, Adelaide, SA 5005, Australia. Tel.: +61 88226 7700; fax: +61 988226 7777.

E-mail address: roger.byard@sa.gov.au (R.W. Byard).

Papers that did not cite one of the three definitions above were placed in either of two categories:

- 4) Those that used idiosyncratic, other or mis-cited definitions, and,
- 5) Those where there was no definition.

3. Results

The 50 papers selected from 2010 to 2011 were drawn from a mixture of original research, case series, meta-analyses and reviews. Forty-five papers either used one or no definition, with five papers citing more than one definition (resulting in 12 definitions in these 5 papers). This gave a total of 57 instances where a definition was either used or not used.

Seattle definition	Four papers, where the definition was not written in the text but was referenced to the original publication. ⁴ (N = 4; 7%).
NICHHD definition	Twenty papers which included: i) 10 papers where the definition was written in the text and referenced to the original publication, ⁵ ii) 5 papers where it was written in the text and referenced to an alternative (but not original) paper, and iii) 5 papers where it was not written in the text but was referenced to the original publication. (N = 20; 35.1%).
San Diego definition	Fifteen papers which included: i) 4 papers where the definition was written in the text and referenced to the original publication, ⁶ ii) 10 papers where it was not written but was referenced to the original publication, and iii) 1 paper where it was written in the text and not referenced. (N = 15; 26.3%).
Idiosyncratic/other/mis-cited	Seven papers which included i) 4 with non-standard definitions written in the text and ii) 3 with no definition and with alternative (not original) references. (N = 7; 12.3%).
Not cited	Eleven papers where no definition was given (N = 11; 19.3%).

4. Discussion

One of the criticisms of SIDS research in the past has been the array of apparently contradictory results that have made determining aetiological pathways difficult. A suggestion in the previous study was that failure to specify a definition of SIDS in over half of the published reports studied may have contributed to this situation.¹ The present study has demonstrated, however, that there has been a significant improvement in the use of definitions, with the percentage of papers either quoting or referencing a standard definition increasing by 26%, from 42 to 68%. While the 1989 NICHHD definition remained the most commonly used definition, there has been a marked increase in reports using the 2004 San Diego definition. This has increased from 10 to 26% in the interval between the studies (Table 1). A small increase from 2 to 7% had also occurred in the use of the 1969 Seattle definition.

Although the usefulness of the definitions relies upon reproducible diagnostic criteria, these have remained elusive, as SIDS is still a 'diagnosis' of exclusion.⁷ Several expert panels have

Table 1

Use of definitions of SIDS in the literature (2005 compared to 2010–2011).

Definition	2005 ¹	2010–2011
1. Seattle	2%	7%
2. Nichd	30%	35.1%
3. San Diego	10%	26.3%
4. Idiosyncratic	16%	12.3%
5. Not cited	42%	19.3%

attempted to deal with the issues of standardising the investigation of SIDS deaths and establishing diagnostic criteria to assist with the evaluation of these cases at autopsy. Their conclusions can be found in the published papers from these groups.^{8,9}

The increased number of authors specifying the definition of SIDS used in their papers has been reflected in the fall from 58 to 32% in the number of papers where either no definition was provided or where an idiosyncratic or mis-cited definition was used. Although this is a welcome trend, it should be noted that nearly one third of papers published on SIDS in peer-reviewed journals that were included in the current study did not have adequate definitions. It would be gratifying if this number had dropped a further 26% should a review of papers be carried out again in another six years. The most appropriate definitions to use are those that have been established as international standards and endorsed by professional organizations. Given that the San Diego definition provides a more detailed list of diagnostic criteria its use is recommended.

Conflict of interest

None.

Funding

None.

Ethical approval

N/A.

References

- Byard RW, Marshall D. An audit of the use of definitions of sudden infant death syndrome (SIDS). *J Forensic Leg Med* 2007;**14**:453–5.
- Mitchell E, Krous HF, Donald T, Byard RW. An analysis of the usefulness of specific stages in the pathological investigation of sudden infant death. *Am J Forensic Med Pathol* 2000;**21**:395–400.
- <http://www.ncbi.nlm.nih.gov/pubmed>. [accessed 10.1.12].
- Beckwith J. *Discussion of terminology and definition of the sudden infant death syndrome*. Seattle: University of Washington Press; 1970.
- Willinger M, James LS, Catz C. Defining the sudden infant death syndrome (SIDS): deliberations of an expert panel convened by the national institute of child health and human development. *Pediatr Pathol* 1991;**11**:677–84.
- Krous HF, Beckwith JB, Byard RW, Rognum TO, Bajanowski T, Corey T, et al. Sudden infant death syndrome (SIDS) and unclassified sudden infant deaths (USID): a definitional and diagnostic approach. *Pediatrics* 2004;**114**:234–8.
- Byard RW. Sudden infant death syndrome - a 'diagnosis' in search of a disease. *J Clin Forensic Med* 1995;**2**:121–8.
- Bajanowski T, Vege A, Byard RW, Krous HF, Arnestad M, Bachs L, et al. Sudden infant death syndrome (SIDS) - Standardized investigations and classification: recommendations. *Forensic Sci Int* 2007;**165**:129–43.
- Rognum TO, Arnestad M, Bajanowski T, Banner J, Blair P, Borthne A, et al. Consensus on diagnostic criteria for the exclusion of SIDS. *Scand J Forensic Sci* 2003;**9**:62–73.